전기진단학적 검사상 다발신경근병증에서 신경얼기병증으로 변화 양상을 보이는 당뇨병성 요천추부 신경근얼기 신경병증: 증례 보고

노규동, 석 현, 김상현, 박민근, 김진영, 이승열

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Diabetic Lumbosacral Radiculoplexus Neuropathy Presenting Progressive Pattern from Polyradiculopathy to Plexopathy on Electrodiagnostic Study: A Case Report

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Lumbosacral radiculoplexus neuropathy (LRPN) can involve various parts of nerve roots and plexus and present with various symptoms and signs. LRPN can be caused by metabolic, inflammatory, traumatic, oncologic and autoimmune disorders. Electrodiagnostic study (EDX) is important to localize the involvement of lumbosacral lesion. In addition to EDX, additional tests including serum, cerebrospinal fluid and magnetic resonance imaging (MRI) are also required to clarify the cause of lesion. In this case, a 63-year-old man with diabetes and history of traffic accident suffered from severe pain and motor weakness on the left lower extremity. Initially, he was diagnosed with lumbar polyradiculopathy based on EDX. However, final diagnosis was confirmed as diabetic LRPN by second EDX and MRI, which means progressive pattern of disease from polyradiculopathy to plexopathy.

Key Words: lumbosacral radiculoplexus neuropathy, electrodiagnostic study, diabetes

Introduction

Symptoms of lumbosacral plexus disorder can appear in a variety of ways on lower torso, pelvis and

legs. Pain, sensory impairment, motor weakness and other symptoms can occur as a result of neuropathy. Accordingly, appropriate treatments for pain and cause of lesion become more important. In the diagnosis of lumbosacral plexus lesion, there are many difficulties to find out not only a range of involvement but also causes of disease. Electrodiagnostic study (EDX) is a useful method for determining the extent of involvement. Lumbosacral plexopathy (LSP) involves two or more peripheral nerves in at least two nerve-root distributions by definition.¹ However, LSP often

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occurs with involvement of the root, plexus and nerve rather than that of only the lumbosacral plexus nerve segment, which cause lumbosacral radiculoplexus neuropathy (LRPN).

We report a unique case confirmed as diabetic LRPN by second EDX and imaging studies, which initially diagnosed as lumbar polyradiculopathy by the first EDX.

Case Report

A 63-year-old man, who was diagnosed with hypertension about 4 years ago and diabetes mellitus about 2 years ago, first visited our outpatient clinic on April 4, 2018 with left lower extremity motor weakness and severe pain (Numeric rating scale, NRS 10/10). He had deep aching and burning pain at first and then gradually began to weaken his thigh and calf muscles. About 4 months ago, he had a history of car accident. At that time, there was no bony problem such as fracture and other definite neuromuscular lesion. However, after that, he had progressive muscle atrophy and pain on left lower extremity. Because of pain and weakness, before visiting our hospital, he had a spinal interven-

tion for 3 times at local pain clinic under suspicion of lumbosacral radiculopathy.

On physical examination, according to the medical research council (MRC) grading system, the motor strength of left hip flexor, knee extensor, ankle dorsiflexor and ankle plantar flexor was 4, 3, 3 and 4, respectively. There was decreased sensation on L4 and L5 dermatome of left lower extremity. Initial motor and sensory nerve conduction study of left lower extremity was within normal limit (Table 1). Needle electromyography (EMG) showed abnormal spontaneous activities such as fibrillation potential and positive sharp wave and decreased interference pattern in left adductor longus, tibialis anterior, peroneus longus, vastus medialis, rectus femoris, gluteus maximus and L3~L5 paraspinalis muscles, concluded as left lumbar polyradiculopathy, L3~5 involved (Table 2). He had mild disc bulging at L3/4, L4/5 levels on magnetic resonance imaging (MRI) taken before 3 months ago, which was considered as the cause of polyradiculopathy.

After taking pain medicine including opioids and pregabalin, pain of left lower extremity had improved by NRS 7 points. Two months later, however, the

Table 1. Nerve Conduction Study

Nerve and sites	Initial (4 months later after onset)			Second (6 months later after onset)		
	Lat, ms	Amp, mV	CV, m/s	Lat, ms	Amp, mV	CV, m/s
Motor conduction study						
Lt peroneal nerve (EDB)	5.00	5.9	41.4	-	-	-
Rt peroneal nerve (EDB)	5.08	5.3	41.2	4.97	4.5	47.0
Lt tibial nerve (AH)	4.67	10.9	42.0	4.92	0.8	35.7
Rt tibial nerve (AH)	4.67	11.1	44.1	4.71	11.7	49.5
Lt femoral nerve (RF)	NT	NT	NT	7.70	0.1	
Rt femoral nerve (RF)	NT	NT	NT	6.38	7.1	
Sensory conduction study						
Lt superficial peroneal nerve	2.36	12.5	42.4	-	-	-
Rt superficial peroneal nerve	2.42	11.3	41.3	2.45	19.2	51.5
Lt sural nerve	2.38	22.5	42.0	-	-	-
Rt sural nerve	2.31	21.9	43.3	2.25	18.3	55.2
Lt LFCN	NT	NT	NT	1.69	16.1	71.0
Rt LFCN	NT	NT	NT	1.65	13.0	72.7
Lt saphenous nerve	NT	NT	NT	6.50	14.0	18.5
Rt saphenous nerve	NT	NT	NT	2.63	41.0	45.6

Rt: right, Lt: left, EDB: extensor digitorum brevis, AH: abductor hallucis, RF: Rectus femoris, Lat: latency, Amp: amplitude, CV: conduction velocity, LFCN: lateral femoral cutaneous nerve, NT: not tested

Table 2. Needle Electromyography

Muscle	Initial (4 months later after onset)			Second (6 months later after onset)		
	Fib	PSW	Recruitment	Fib	PSW	Recruitment
Lt Adductor longus	1+	1+	3/5	1+	1+	1/5
Lt Tibialis anterior	1+	1+	1/5	3+	3+	0/5
Lt Tibialis posterior	–	–	3/5	2+	2+	0/5
Lt Peroneus longus	1+	1+	3/5	3+	3+	0/5
Lt EHL	–	–	3/5	–	–	3/5
Lt Gastrocnemius	–	–	3/5	2+	2+	0/5
Lt Vastus medialis	1+	1+	2/5	2+	2+	2/5
Lt Iliopsoas	–	–	3/5	–	–	4/5
Lt Rectus femoris	2+	2+	2/5	1+	1+	3/5
Lt Semitendinosus	NT	NT		2+	2+	2/5
Lt BFL	NT	NT		–	–	2/5
Lt Gluteus medius	2+	2+	4/5	2+	2+	2/5
Lt Gluteus maximus	–	–	4/5	1+	1+	2/5
Lt L3 paraspinalis	1+	1+		–	–	
Lt L4 paraspinalis	1+	1+		–	–	
Lt L5 paraspinalis	1+	1+		–	–	
Lt S1 paraspinalis	–	–		–	–	

Lt: left, Fib: fibrillation potential, PSW: positive sharp wave, EHL: Extensor hallucis longus, BFL: Biceps femoris long head, NT: not tested

motor weakness of left lower extremity had gotten a little worse. According to motor MRC score, the motor strength of left hip flexor, knee extensor, ankle dorsiflexor and ankle plantar flexor was 3, 3, 1 and 1, respectively. There was atrophy in left thigh muscles. Circumference differences between two legs at points located 10 cm and 15 cm proximally from the base of patella were 3 cm and 2.5 cm, respectively. He was hospitalized and had several evaluations. For rule out other causes, serum studies including tumor markers and brain computed tomography (CT) were conducted, which showed no abnormal results. Blood glucose level was well controlled with oral medication. In addition, cerebrospinal fluid (CSF) study for differentiating inflammatory neuropathy showed normal white blood cell and protein level. In follow up EDX, sensory nerve conduction study showed absent response of left sural and superficial peroneal nerves; delayed latency, decreased amplitude and slowed conduction velocity of left saphenous nerve; normal response of left lateral femoral cutaneous nerve. Motor nerve conduction study showed absent response of left peroneal nerve; decreased amplitude and slowed conduction velocity of left tibial and sciatic nerves; delayed la-

tency and decreased amplitude of left femoral nerve (Table 1). Needle EMG showed abnormal spontaneous activity in most of left lower extremity except for hip flexor and paraspinalis muscles (Table 2). Spine and pelvis MRI with enhancement showed disc bulging in L4-5 with annular fissure and diffuse thickening and enhancement of left sacral plexus and sciatic nerve (Fig. 1, 2). Increased T2 signal and enhancement of left sacral plexus and sciatic nerve indicated the inflammation of lumbosacral plexus. Positron emission tomography-computed tomography (PET CT) for excluding cancer showed focal increased uptake along the left sciatic nerve at the posterior aspect of acetabulum (Fig. 3). However, because there was no evidence of malignancy or metastasis, inflammation was considered as the cause of increased uptake in PET CT. Combining all the results including progressive pattern from polyradiculopathy to plexopathy on EDX, and MRI and PET CT findings, the patient was finally diagnosed with diabetic lumbosacral radiculoplexus neuropathy. One year after the onset of symptoms, his pain showed some improvement by pregabalin and opioids, but motor weakness persisted.

Fig. 1. Lumbar spine MRI T2 weighted image. Sagittal view (A) and Axial view (B). Disc bulging in L4-5 with annular fissure L4/5 is noted (arrows).

Fig. 2. Pelvic MRI T2 weighted gadolinium enhanced fat suppression image (A-C). Diffuse thickening of left sacral plexus and sciatic nerve with enhancement is noted (arrows).

Fig. 3. PET CT. Torso (A) and axial fusion image (B). Focal increased uptake is noted along the left sciatic nerve at the posterior aspect of acetabulum (arrows).

Discussion

This case is about the patient who had a history of diabetes and trauma (traffic accident) and was presented as lumbar polyradiculopathy by initial EDX study at 4 months after symptom onset, but finally diagnosed with diabetic lumbosacral radiculoplexus neuropathy based on results of second EDX testing and imaging studies.

LRPN is caused by various disease processes, such as neoplasm, infection, trauma, radiation treatment of pelvic neoplasm, hematoma, ischemia, inflammation, infiltration and idiopathic causes. In our case, patient had diabetes for 2 years and car accident about 4 months ago. Although there was trauma history, traumatic neuropathy by car accident is less likely due to onset of symptoms that progressed after trauma over 4 months and MRI findings showing no other musculoskeletal lesion. Plexus lesion caused by inflammatory/microvasculitis shows asymmetric lower limb weakness, sensory loss and paresthesia with spontaneous pain of acute or subacute onset. The causes of these include diabetic LRPN, nondiabetic LRPN and postsurgical inflammatory neuropathy. Patients with diabetic LRPN usually present with acute to subacute onset of pain with motor weakness and has relatively well-controlled blood glucose level. Our patient also controlled his blood glucose well without insulin and had a subacute pain with motor weakness on unilateral lower extremity, which means that diabetic LRPN is more likely.

Diabetic LRPN is caused by inflammation and microvasculitis, which can be seen as findings of ischemic injury, perivascular inflammation, and microvasculitis in nerve biopsies.² It can involve nerve roots, peripheral nerve and plexus, causing neuropathic pain followed by motor weakness and sensory loss. Diabetic LRPN commonly affects the proximal thigh and can also involve sciatic distribution. However, involvement of sciatic distribution is less common in the early phase of disease compared to the proximal

weakness. Later course of disease, pain and weakness may spread to distal leg segments.³ Most patients have improvement within 1 to 3 years after onset, but many have residual neurologic deficits at recovery.⁴

LRPN is suspected through clinical history and neurologic examination and requires many tests such as blood, EDX, CSF, MRI and PET CT for diagnosis. Among the many tests, accurate localization of lumbosacral plexus lesion requires an EDX.⁵ If they show unilateral involvement, comparison with contralateral side could be useful.⁶ In case of pure LSP, there should be no involvement of paraspinal muscles. In EDX, LSP is also distinguished from common lumbosacral radiculopathy which usually showed normal sensory nerve action potential (SNAP) and denervation of paraspinal muscle. However, they often occur with involvements of root, plexus, and nerve, causing LRPN.⁷ Initial EDX of this case showed denervation potentials of lumbosacral paraspinalis muscles which means involvement of nerve roots. Moreover, sensory nerve conduction study was normal, which means that initial lesion is proximal to the dorsal root ganglion. Without compressive lesion, radiculopathy can be caused by “chemical radiculitis” associated with leakage of chemical mediators from disc into epidural space through annular tear.⁸ However, there was only annular fissure in L4~5 level in spine MRI, which was insufficient to be a main cause of polyradiculopathy. In second EDX, there was no denervation of lumbosacral paraspinalis muscles and only lesion of lumbosacral plexus, which can be interpreted as a progression of lesion.

To diagnose a cause of lumbosacral lesion, several tests are required as well as EDX. In inflammatory LRPN, CSF study is important, showing isolated elevating of protein without elevation of white cells.⁷ If a patient is suspected of neoplasm, traumatic injury or compression of lumbosacral plexus, MRI may be considered among the neuroimaging study.^{9,10} Diabetic LRPN and postsurgical inflammatory neuropathy causing LRPN or LSP could show abnormal findings

with increased T2 signal or normal finding. In addition, gadolinium-enhanced MRI could show inflammation of nerve. Although CSF study was normal in our case, MRI findings of increased T2 signal and enhancement are also evidence of diabetic LRPN. Increased uptake in PET CT which is important in case of suspecting malignancy also means inflammatory cause.¹¹ In addition, Nerve biopsy can show ischemic injury from microvasculitis which occur in inflammatory neuropathy.⁷

In this case, considering clinical manifestation and results of diagnostic tests, diabetic LRPN which sequentially involved lumbosacral root and plexus is more likely than traumatic lumbosacral plexopathy. To our knowledge, this is the first report of presenting alteration from radiculopathy to plexopathy pattern on EDX in diabetic LRPN, which can be confused as misdiagnosis or new overlying disease.

In conclusion, diabetic LRPN can manifest as a variety of symptoms and involve various regions. Moreover, the course of disease also can change with time, which makes difficult to diagnosis. Comprehensive workups in the progress of a disease are important for accurate diagnosis and proper management.

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References

1. Laughlin RS, Dyck PJB, PM, Clinics R: Electrodiagnostic testing in lumbosacral plexopathies. 2013; 24: 93-105
2. Tracy JA, Engelstad JK, Dyck PJ: Microvasculitis in diabetic lumbosacral radiculoplexus neuropathy. J Clin Neuromuscul Dis 2009; 11: 44-48
3. Bhanushali MJ, Muley SA: Diabetic and non-diabetic lumbosacral radiculoplexus neuropathy. Neurol India 2008; 56: 420-425
4. Dyck PJ, Norell JE, Dyck PJ: Non-diabetic lumbosacral radiculoplexus neuropathy: natural history, outcome and comparison with the diabetic variety. Brain 2001; 124: 1197-1207
5. Kimura J: Electrodiagnosis in diseases of nerve and muscle: principles and practice: Oxford university press, 2001.
6. Barr K: Electrodiagnosis of lumbar radiculopathy. Phys Med Rehabil Clin N Am 2013; 24: 79-91
7. Dyck PJ, Thaisetthawatkul P: Lumbosacral plexopathy. Continuum (Minneap Minn) 2014; 20: 1343-1358
8. Peng B, Wu W, Li Z, Guo J, Wang X: Chemical radiculitis. Pain 2007; 127: 11-16.
9. Maravilla KR, Bowen BC: Imaging of the peripheral nervous system: evaluation of peripheral neuropathy and plexopathy. AJNR Am J Neuroradiol 1998; 19: 1011-1023
10. Taylor BV, Kimmel DW, Krecke KN, Cascino TL: Magnetic resonance imaging in cancer-related lumbosacral plexopathy. Mayo Clin Proc 1997; 72: 823-829
11. Vaidyanathan S, Patel CN, Scarsbrook AF, Chowdhury FU: FDG PET/CT in infection and inflammation--current and emerging clinical applications. Clin Radiol 2015; 70: 787-800